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Centre-level variation in treatment and outcomes and predictors of outcomes in 5-year-old children with non-syndromic unilateral cleft lip treated within a centralized service: the Cleft Care UK study. Part 6: summary and implications

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Structured Abstract

Objectives: To summarize and discuss centre-level variation across a range of treatment and outcome measures and examine individual and ecological determinants of outcome in children in Cleft Care UK (CCUK).

Setting and sample population: Two hundred and sixty-eight 5-year-old British children with non-syndromic unilateral cleft lip and palate (UCLP) recruited to CCUK and treated within a centralized service.

Materials and Methods: Children had a range of treatment and outcome measures collected at a 5-year audit clinic. These outcomes included dento-alveolar arch relationships from study models, measures of facial appearance from cropped photographs, hearing loss from audiological assessment, speech from speech recordings, self-confidence and strengths and difficulties from parental self-report. Data were collected on educational attainment at age 7 using record linkage. Centre variation was examined using hierarchical regression and associations between variables were examined using logistic or poisson regression.

Results: There was centre-level variation for some treatments (early grommet placement, fitting of hearing aids, fluoride treatment, secondary speech surgery and treatment for cleft speech characteristics) and for some outcomes (intelligibility of speech). Hearing loss was associated with a higher risk of poor speech while speech therapy was associated with a lower risk of poor speech. Children had high levels of caries but levels of preventative treatment (fluoride varnish and tablets) were low.

Conclusions: Further improvements to and monitoring of the current centralized model of care are required to ensure the best outcomes for all children with cleft lip and palate.

KEYWORDS

centralization, cleft lip, cleft palate, outcomes, variation

1 | INTRODUCTION

In this supplement, we report further on a UK-wide cross-sectional survey of 5-year-old children with unilateral cleft lip and palate conducted between January 2011 and December 2012—Cleft Care UK (CCUK)¹⁻⁵ that we have described previously.⁶⁻¹¹ We have already shown that some outcomes (such as facial growth, speech and parental report of self-confidence) have improved following centralization^{7,9,10} while some outcomes (such as oral health and hearing) have not changed.⁸ But a proportion of children still have poor outcomes within this centralized multidisciplinary service¹² (even for outcomes that have improved). Our previous supplement focused on comparisons with a previous cross-sectional study conducted 15 years previously.¹³ In this supplement, we describe treatment and outcomes in more detail. For example, we present new summary articulation and structural scores for speech⁴ and strengths and difficulties scores for psychosocial outcomes,⁵ explore centre-level variation in treatment and outcome and examine individual and ecological determinants of outcome and sequelae of poor outcome within a centralized service.

2 | CENTRE-LEVEL VARIATION

We used hierarchical regression to predict overall and centre-level means, and to estimate the variance partition coefficient (VPC)—a measure of the amount of variation in treatment or outcome explained by the centre.¹ We have summarized the centre-level variation for all treatments and outcomes in Table 1. We found centre-level variation for some treatments such as early grommet placement (VPC 18%,

$P < .001$), fitting of hearing aids (VPC 8%, $P = .03$),² fluoride treatment (eg, VPC for fluoride tablets: 57%, $P < .001$),³ secondary speech surgery (VPC 15% $P < .001$) and treatment for cleft speech characteristics (VPC 9% $P = .006$).⁴ There was also centre-level variation for some outcomes such as intelligibility of speech (VPC 13%, $P = .001$) with the centres with the best results having half the number of children with poor intelligibility of speech.⁴ Although our nationwide study was large (for a study of children with cleft lip and palate), had a good response rate and a series of validated measures of key outcomes with enough precision to demonstrate improvements over time, it had limited power to detect centre-level variation. Even so we were able to show clinically meaningful levels of variation in some treatments and outcomes across this centralized service but we were unable to explore centre-level characteristics such as size and time since centralization. Further, it may be that for some outcomes such as dento-alveolar arch relationships that the key unit of variation is not the centre but the surgeon and that this explains the lack of centre-level variation observed for some outcomes. While centre-level variation does not explain why outcomes vary, its presence does suggest that the observed variation may be modifiable.

3 | OUTCOMES THAT IMPROVED WITH CENTRALIZATION

Dento-facial outcomes and self-reported self-confidence have improved with centralization,^{7,10} but we found no evidence of centre-level variation for either outcome. For dento-facial outcomes, this lack of variation may be because there is little variation in outcome within a centralized service.¹ However, it could reflect the difficulty in measuring

TABLE 1 Centre-level variation in treatments and outcomes

Domain	Outcome variable	% variance explained	P-value	Treatment variable	% variance explained	P-value
Facial growth	Good (1 or 2)	4	.6			
	Poor (4 or 5)	3	.9			
Facial appearance	Good (1 or 2)	2	.9			
	Poor (4 or 5)	5	.9			
Audiology	Best hearing levels	4	.3	Grommets	3	.9
	Worst hearing levels	2	.9	Grommet(s) at palate closure	18	<.001
	History of hearing loss/OME	3	.9	Fitted hearing aid	8	.03
	Current hearing problem (best ear >20 dB)	2	.5			
Oral health	Caries	2	.3	Fluoride tablets	57	<.001
	Rampant decay	2	.9	Fluoride varnish	9	.02
				Fluoride in toothpaste (1000 ppm+)	3	<.001
Speech	Articulation score	5	.3	Secondary speech surgery	15	<.001
	Structural score	5	.08	History of treatment for CSC	9	.006
	Poor intelligibility	13	<.001	Treatment required but not in therapy	8	.10
Psychology	Self-confidence	1	.9			
	Total difficulties	3	.9			

CSC, cleft speech characteristics; OME, Otitis media with effusion. Adjusted for age and sex.

TABLE 2 Individual- and area-level associations with key outcomes and exposures

Association examined	Factors	Effect size
Sequela of OME and hearing loss	Speech poor intelligibility ↓	OR=2.9 (95% CI 1.4 to 5.8)
	Speaking and listening ↓	OR=1.8 (95% CI 0.8 to 4.0)
Predictors of oral health	Use of fluoride tablets ↑	RR 1.7 (95 CI 1.3 to 2.3)
	Use of fluoride varnish ↑	RR 1.3 (95 CI 1.0 to 1.7)
Predictors of poor speech	Treatment for speech ↓	OR=0.1 (95% CI 0.0 to 0.4)
	Parental concern ↑	OR=13.2 (95% CI 4.9 to 35.1)

appearance reliably,⁷ the limited resolution of values using current scores for dento-alveolar arch relationships to discriminate between children, a lack of power to detect small or modest variation between sites or the fact that the key unit of comparison should have been the individual surgeon rather than the larger multidisciplinary centre. We extended the range of behavioural outcomes to include the strengths and difficulties questionnaire.¹⁴ We found that children had a higher hyperactive difficulties score than the general population but that there was no evidence of centre-level variation for behaviour.⁵ For speech outcomes, though, we found that they have improved with centralization we also found evidence of centre variation in both therapy and outcomes.⁴ Further we found evidence that speech therapy was effective and that parental concern is a good predictor of poor speech.⁴

4 | OUTCOMES THAT WERE UNCHANGED BY CENTRALIZATION

For oral health, we have previously reported that there was no improvement in outcome with centralization⁸ and we found no evidence of centre-level variation in outcomes.³ We found that children had high levels of caries, rampant caries and developmental enamel defects but that levels of preventative treatment were low with fluoride tablets and varnish probably being used after caries have developed rather than before.³ Moreover, there was evidence of variation in these fluoride treatments which were more commonly used in centres that had a paediatric dentist. For hearing, there was no improvement with centralization⁸ but although there was no centre-level variation in outcomes, there was substantial variation in treatment between centres.² We also found that a history of otitis media with effusion and hearing loss was associated with poor intelligibility of speech at age 5 and potential aspects of educational attainment at age 7.

5 | IMPLICATIONS FOR PRACTICE

Our previous findings showing an improvement in outcomes support the value of introducing and maintaining a centralized multidisciplinary service. We did not find any centre-level variation in dento-facial

outcomes so our results do not help to explain why a proportion of children have poor dento-facial outcomes within a centralized service. For treatments or outcomes where there has been no improvement following centralization or where there is centre-level variation in treatment or outcome, centres need to review and justify variations in practice. The treatments used and the care provided should be the focus of audits to identify areas where improvements can be made. For hearing, the variations in treatment and associations with outcomes suggest that treatment for hearing loss can be improved. There is a need for closer collaboration between speech and language therapists and audiologists/ENT surgeons managing the care of children with cleft lip and palate to ensure hearing loss is actively managed, which includes therapeutic support for speech, listening and communication development. For oral health, our data suggest that centre-wide and national improvements in service provision are required. Paediatric dentists are now present in all but one centre which was not the case when the centralization process began, but this provision has not been consistent and is very recent in some centres. We suggest that an integrated intensive programme of preventative oral health care is required for children with cleft lip and palate. Earlier work on this issue in Ireland suggests improvements would be seen within 5 years of implementing this approach.¹⁵ Our findings of centre-level variation in treatment for poor speech and speech outcomes support the early identification and management of fistulae, effective management of velopharyngeal insufficiency and hearing impairment, and most importantly access to speech intervention in the preschool years. There is a need for equitable and adequate provision of speech and language therapists and closer working relationships with audiology and ENT services to encourage the optimal development of communication skills in children with cleft lip and palate.

6 | IMPLICATIONS FOR RESEARCH

Further cross-sectional studies are required to monitor changes in treatment and outcome that are large enough to have adequate power to detect clinically important temporal and centre-level differences in treatment and outcomes. They also need more detailed measures of centre-level attributes (such as details of individual surgeon characteristics), outcomes and additional measures of levels of language development and intelligence so that the determinants of key outcomes such as speech can be better described. Longitudinal studies could help to describe the impact of treatments (such as hearing aids, speech and language therapy or fluoride varnish) and other factors on outcome. Where possible, trials should be considered to identify effective (and ineffective) treatments as at present there is no robust trial evidence to guide practice in a number of areas.¹⁵⁻¹⁷

7 | CONCLUSIONS

Centralization as we have already shown improves outcome but it is not a panacea. There are variations in treatment and outcome within



this centralized service. Although interestingly for the outcomes that have not improved and/or where there is centre-level variation, the service is often either not integrated with the multidisciplinary centre or is provided in the community rather than by staff at the centre. Further improvements to and monitoring of the current centralized model of care are required to ensure the best outcomes for all children with cleft lip and palate.

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